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A case of extensive bilateral conjunctival habronemiasis in a Connemara gelding

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Summary: Habronemiasis is a parasitic disease caused by one of three species of nematodes. Adult stages of these worms live in the equine stomach. Larval stages can cause cutaneous forms of habronemiasis, muco-cutaneous forms on the ocular or genital mucosa, as well as pulmonary and erratic forms. Although most prevalent in tropical and subtropical areas, it is distributed worldwide. However, the prevalence of the cutaneous and muco-cutaneous forms is lacking in Western Europe. This report presents a rare form of habronemiasis in the periocular region of a 10 year old Connemara pony in Western France. The pony was presented for evaluation and treatment of his right eye, showing signs of pain for 10 days. Despite previous treatments, no improvement had been observed. The pony had been treated the previous year for periocular habronemiasis with corticosteroid injections. Clinical examination revealed marked blepharospasm with moderate epiphora on the right eye. Yellow granules, reaching 1.5 mm in diameter, could be observed in the medial canthus, close to the lacrimal puncta. The lower eyelid and medial canthus were thickened and numerous hard similar granules could be palpated beneath under the palpebral conjunctiva. Fluorescein dye showed a 2 cm large stromal ulcer facing the previously described granules. Jones test was negative. On the left eye also, the lower eyelid was thickened and subconjonctival granules could be palpated, giving this pony an appearance of bilateral ectropion. A diagnosis of unilateral corneal ulcer and bilateral ectropion likely due to chronic habronemiasis-related sulphur-like aranules was made. On the right eye, the visible granules were removed under standing sedation and medical treatment was initiated, allowing corneal healing in two weeks time. Two months later the pony was presented for another corneal ulcer on his right eye. The latter was medically treated and surgical intervention was planned in order to remove the sulphur-like granules on both eyes. Under general anaesthesia, the palpebral and nictitating membrane conjunctiva were incised where the granules could be palpated. They were dissected free. Larger incisions were sutured whereas small ones were left open to heal by secondary intention. Large granules were removed from the right medial canthus through a V-shaped incision that was subsequently closed in two layers. On the right eye, a canaliculosinostomy was then performed. A few granules were fixed in buffered formalin for histopathological analysis. Histopathology revealed a bilateral multifocal eosinophilic granulomatous conjunctivitis, of chronic course and marked intensity, associated with intralesional larval residues, compatible with spiruroid nematode larvae of Habronema sp. Topical antibiotics were given for 10 days, followed by topical steroids for 7 days. After tube removal, 4 weeks postoperatively, the pony showed neither swelling nor discharge on both eyes with no visible ectropion. No further corneal ulceration was detected for at least a year postoperatively. This pony is the first reported case of cutaneo-mucous habronemiasis in France. It is unique by the large number of dense fibrous granules causing ectropion and corneal ulcerations.

Keywords: horse, ophthalmology, habronema, keratitis, canaliculosinostomy

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Introduction

Habronemiasis, also called habronemiosis or habronematidosis is a parasitic disease caused by either *Habronema muscae*, *Habronema microstoma* (synonymous *H. majus*) or *Draschia megastoma*. House- and stable-flies act as intermediate hosts. Adult stages of these nematode species live in the stomach of domestic and wild equids. Larval stages can be responsible for cutaneous forms of habronemiasis or summer sores, muco-cutaneous forms on the ocular or genital mucosa, as well as pulmonary and erratic forms (*Barlaam* et al. 2020). Although most prevalent in tropical and subtropical areas, it is distributed worldwide. In Western Europe, gastric

habronemiasis has been reported in 1.1% of horses at necropsy in Sweden (Hoglund et al. 1997), 4.3% in The Netherlands (Borgsteede and van Beek 1998), 8.5% in France (Collobert-Laugier et al. 2000), 17% in Belgium (Pecheur et al. 1979), and 33% in Germany (Rehbein et al. 2013). However, the prevalence of the cutaneous and muco-cutaneous forms is lacking in these countries with descriptions only occurring through case reports (Gasthuys et al. 2004, Read et al. 1996, Verhaar et al. 2018). Its presence has also anecdotally been confirmed by equine practitioners in Southern France (Bourdoiseau and Pin 2018). This report presents a rare form of habronemiasis in the periocular region of a Connemara pony in Western France.

Case presentation

A 10 year old grey Connemara gelding, bred in Western France, was presented in May 2020 for evaluation and treatment of his right eye, showing signs of pain for 10 days. Despite previous treatments with topical atropine, antibiotics and anti-inflammatory drugs, no improvement had been observed. He had reportedly been treated the previous year for periocular habronemiasis on both eyes with corticosteroid injections. He had been receiving a macrocyclic lactone (either moxidectine or ivermectine) three times a year since he belonged to his current owner, i.e. for 4 years.

Clinical examination revealed marked blepharospasm with moderate epiphora on the right eye. After sedation, an auriculopalpebral nerve block (Manning and St Clair 1976) was performed. Yellow granules could be observed in the medial canthus, around the dorsal and ventral lacrimal puncta. These granules were reaching 1.5 mm in diameter at some places. The lower eyelid and medial canthus were thickened and numerous hard structures, believed to be similar granules, were palpated beneath the palpebral conjunctiva. Fluorescein dye showed a 2 cm large stromal ulcer in the nasal part of the right eye, facing the previously described granules. Jones test was negative (Featherston and Heinrich 2013). Moderate diffuse corneal oedema, as well as a few vessels originating from the ventral limbus, were visible around the ulcer (Figure 1).

On the left eye, the lower eyelid was also thickened and sub-conjonctival granules could similarly be palpated. These abnormalities gave the pony an appearance of suffering from ectropion on both sides. A diagnosis of unilateral stromal ulcer and bilateral ectropion likely due to chronic habronemiasis-related sulphur granules was made.

On the right eye, the visible granules were removed under standing sedation and local anaesthesia using Bishop-Harmon tissue forceps and a subpalpebral lavage (SPL) system was inserted. The pony was treated aggressively in-hospital for a week using topical tobramycin, rifampicin, itraconazole and autologous serum. Systemic flunixin meglumine was also given and progressively tapered off according to the pony's comfort level. Treatments were continued at home for a week before removing the SPL when the corneal ulcer was healed. Removal of the granules located under the conjunctiva of both eyes was recommended to prevent further complications.

Two months later the pony was presented again to the clinic for another superficial corneal ulcer on the right eye. The latter was medically treated and surgical intervention was planned in order to remove the sulphur-like granules on both eyes. Preoperative examination showed a large ulcerated granuloma on top of the right ventral lacrimal punctum. Jones test was again negative.

Under general anaesthesia, using a standard protocol, in dorsal recumbency with the neck tilted to the left, the right eye was operated first.

After routine preparation for surgery, the inferior palpebral conjunctiva was incised where the granules could be palpat-

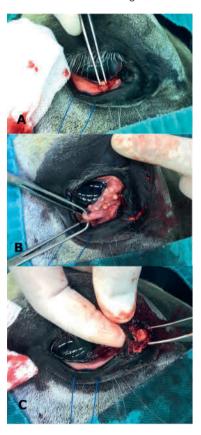


Fig. 2 Surgical removal of the subconjunctivally located sulphur-yellow granules in the lower eyelid (A), in the third eyelid (B) and in the medial canthus (C). | Chirurgische Entfernung des subkonjunktival gelegenen Schwefelgranulats im unteren Augenlid (A), im dritten Augenlid (B) und im medialen Canthus (C).

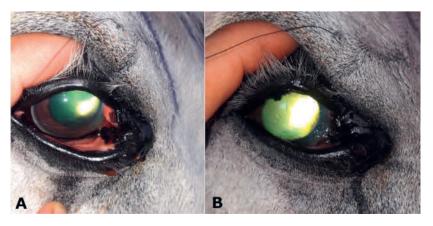


Fig. 1 Right eye of a 10-year old Connemara gelding upon admission. Notice the thickened appearance of the lower eyelid and medial canthus with subsequent moderate ectropion (A). A large stromal ulcer is visible nasally as well as moderate diffuse corneal oedema around it and vessels at the ventral limbus (B). | Rechtes Auge eines 10-jährigen Connemara-Wallachs bei Aufnahme. Schwellung des unteren Augenlids und des medialen Canthus mit moderatem Ektropium (A). Ausgedehntes stromales Hornhautülkus nasal sowie ein mäßiges diffuses Hornhautödem am ventralen Limbus (B).

ed using a #64 Beaver blade (Figure 2). The granules were dissected free using Castroviejo scissors. Larger incisions were closed using a simple continuous pattern of 7–0 USP (0.5 metric) polyglactin 910 whereas small ones were left open to heal by secondary intention. The right nictitating membrane, when retracted caudally, showed numerous subconjunctival granules. These were removed similarly. Large granules could also be palpated in the medial canthus. A V-shaped incision was performed in the canthal skin and granules were dissected. After lavaging the wound, subcutaneous tissues and skin were closed in a simple continuous pattern of 4–0 USP (1.5 metric) polyglactin 910. A canaliculosinostomy was then realized, combining features described by *Brink* and *Schumacher* 2016 and *Robinson* et al. 2016.

The pony's neck was then tilted to the right and the left sub-conjunctival granules were removed in a similar fashion. A few granules were fixed in 10% neutral buffered formalin for histopathological analysis (Figure 3).

Recovery was uneventful. Perioperative treatments included procaine penicillin (22000 IU, every 12 hours) for 3 days and flunixin meglumine (1.1 mg/kg, every 24 hours for 3 days then 0.5 mg/kg for 3 days). The pony also received topical tobramycin 3 times a day for 10 days, followed by topical steroids, 3 times a day for 7 days. Recommendations were given to remove the canaliculosinostomy tube 4 weeks postoperatively.

Histopathology revealed multiple coalescing eosinophilic granulomas. These were composed of a necrotic to liquefied acidophilic core with degenerated eosinophils, surrounded by a dense fibrous tissue containing eosinophils and mac-



Fig. 3 Sulphur-yellow granules surgically removed from the left (left swab) and right (right swab) palpebral conjunctiva on a 10 year old Connemara gelding. | Schwefel-gelbe Granula aus der Bindehaut infolge chronischer Habronema-Infektion bei einem 10 Jahre alten Connemara-Wallach.

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intraläsionalen Vorhandensein von Larvenresten assoziiert ist.

rophages. For 4 out of 38 observed granulomas, it was associated with intralesional larval residues, 50 to $80\mu m$ long, compatible with spiruroid larvae of *Habronema* sp (Figure 4). After tube removal, the pony showed neither swelling nor discharge on both eyes with no visible ectropion. No further corneal ulceration was detected for at least a year postoperatively.

The owner was very satisfied about the cosmetic outcome after surgical treatment (Figure 5).

Discussion

This report presents a rare form of habronemiasis in the periocular region of a Connemara pony that was successfully treated by surgery.

It is believed that habronema larvae live for less than a month in cutaneous tissues and that dying of these larvae, rather than the living parasite itself, causes a hypersensitivity reaction leading to granuloma formation, necrosis and calcification, in the form of the pathognomonic sulphur granules, where it occurs (Verhaar et al. 2018, Waddell 1969). These reactions are reportedly common in the medial canthus, caruncle or proximal lacrimal system (Barlaam et al. 2020, Carr 2019, Giuliano 2017, Pusterla et al. 2003), can be responsible for conjunctivitis, blepharitis, ectropion or chemosis (Barlaam et al. 2020, Pusterla et al. 2003), and can lead to erosion, ulceration and even abscessation of the apposing cornea (Bourdoiseau and Pin 2018, Brooks et al. 2017, Read et al. 1996). However, we are not aware of similar cases where the sulphur-yellow granules were found in such large numbers as to cause bilateral ectropion and corneal ulcerations to the extent observed here.

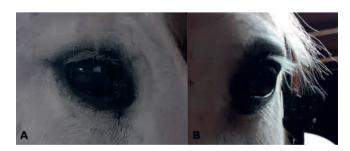


Fig. 5 Appearance of the right (A) and left (B) eyes 3 months postoperatively. Notice the absence of ectropion and minimal swelling on the lower eyelid and medial canthus regions. | Aussehen des rechten (A) und linken (B) Auges 3 Monate postoperativ. Beachten Sie das Fehlen von Ektropium und die minimale Schwellung im unteren Augenlid und im medialen Canthusbereich.

Fig. 4 Histopathological images of one of the submitted granules. Hematoxylin-eosin-saffron stain. A: Magnification 50; It features a chronic eosinophilic granuloma centered on larval residues, associated with a dense fibrous tissue. B: Magnification 200. Intralesional larval residues, 50 to 80 µm long, compatible with spiruroid larvae of Habronema sp. | Histopathologisches Bild eines der Granula. Hämatoxylin-Eosin-Safran-Färbung, die ein multifokales eosinophiles granulomatöses Infiltrat zeigt, das mit dem

The lesions observed in this case suggest a previous severe periocular larval infestation despite apparent regular deworming using macrocyclic lactones such as ivermectin or moxidectin. Alternatively, some degree of anthelminthic resistance has also been suggested recently in habronema species (Kaplan and Vidyashankar 2012, Verhaar et al. 2018). Interestingly, grey horses, such as this, were previously identified at risk for habronemiasis, likely because of the high contrast occurring in the periocular region, attracting houseflies (Howard and Wall 1998, Pusterla et al. 2003).

Recommended treatments for habronemiasis typically involves ivermectin administration, debulking of the lesion, anti-inflammatory therapies, systemically, topically or via injections, and fly control strategies (Giuliano 2017, Pusterla et al. 2003). Nevertheless, because of the chronic nature of the lesions, the fact they were creating corneal ulcers, and the large number of granules, surgical intervention was deemed mandatory in this case, as suggested previously (Bourdoiseau and Pin 2018, Gasthuys et al. 2004, Read et al. 1996). The canaliculosinostomy performed on this case was used in order to alleviate the nasolacrimal duct obstruction caused by the sulphur granules and granulomatous lesions in the medial canthus. The negative Jones test was used, as previously described, as a determining factor when planning the surgical procedure (Robinson et al. 2016). This approach resolved the previously observed epiphora with minimal blemish (Brink and Schumacher 2016).

The histopathological findings described here were typical of spiruridae larval infections (Barlaam et al. 2020, Verhaar et al. 2018). Interestingly, no evidence of mineralization was observed on the submitted samples but, instead, the granulomas were embedded in a dense fibrous tissue. Even though visible in only 44% biopsy specimens in one report (Pusterla et al. 2003), sections of nematodes were observed in this case. Yet, definitive identification of the species involved was not possible. A PCR test was recently developed for detection and identification of Habronema microstoma and Habronema muscae DNA but was not used in this case (Giangaspero et al. 2010).

This case is, as far as the authors are aware, the first reported case of cutaneo-mucous habronemiasis in France. The possible increased prevalence of habronemiasis in Western Europe, as recently mentioned by Verhaar et al. (2018), could be explained by several factors such as climate change, importation of infested horses, selective anthelmintic treatment policy currently recommended, as well as some degree of anthelminthic resistance (Bourdoiseau and Pin 2018, Giuliano 2017, Kaplan and Vidyashankar 2012, Verhaar et al. 2018). As a conclusion, this case is unique by the large number of dense fibrous granules causing ectropion and corneal ulcerations in a pony located in France. Cutaneo-mucous habronemiasis and its consequences should be included in the differential diagnosis of eyelid deformities with associated corneal ulcerations.

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Conflict of Interest Statement

The authors declare that this study was conducted in the absence of any commercial or financial relationships that could be seen as a potential conflict of interest.

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